International Journal of Contemporary Pediatrics
Jain N et al. Int J Contemp Pediatr. 2025 Nov;12(11):1854-1857
http://www.ijpediatrics.com
pISSN 2349-3283 | eISSN 2349-3291

DOI: https://dx.doi.org/10.18203/2349-3291.ijcp20253302

Case Report

Prepubertal-type mature cystic teratoma of the testis in an infant: a rare benign entity with excellent prognosis-managed with testis sparing approach

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Received: 16 September 2025
Revised: 30 September 2025
Accepted: 01 October 2025

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ABSTRACT

Testicular tumors in infants are rare, accounting for only 1-2% of pediatric solid tumors. Among these, teratomas are the most common type, particularly in the prepubertal age group. Unlike their post-pubertal counterparts, prepubertal-type mature teratomas are usually benign, lack malignant potential, and are remotely associated with germ cell neoplasia *in situ* (GCNIS). While mature teratomas are typically found in the abdomen, intratesticular prepubertal-type teratomas in infants are infrequent. The present study describes the case of an infant with an intratesticular mature teratoma. Five months old infant presented with painless right sided scrotal swelling detected incidentally. Scrotal ultrasound revealed a large anechoic cystic lesion measuring 28×18 mm with no septation, calcification, or solid component. Blood parameters including tumor markers viz. alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin (β-hCG) were within normal range for age. Surgical tumor enucleation (testis sparing surgery) was performed, and the histopathological examination revealed a benign, prepubertal-type teratoma composed entirely of mature elements. Based on the findings diagnosis of prepubertal-type mature teratoma (MT) was thus made. Child was later was kept under observation in follow-up. Surgical intervention is commonly used for the management of benign testicular tumors in pediatric patients, including prepubertal teratomas. Indeed, testis-sparing surgery is a recommended option in prepubertal age patients with such a presentation of testicular mass with a benign ultrasonography finding like homogeneous, unilocular, non-septated cyst without any internal echoes, no calcification, no solid component and negative serum markers.

Keywords: Germ cell tumors, Scrotal swelling, Mature teratoma, Alpha-fetoprotein, β-hCG

INTRODUCTION

Prepubertal testicular tumors are rare, accounting for 1%-2% of all pediatric tumors, with an incidence of 0.5-2.0/100,000 children.[1] In newborns and children, pure yolk sac tumors and teratomas constitute the majority of cases, while seminomas and embryonal carcinomas are uncommon.[2-4] Teratomas, the most common tumors found in infants, develop from germ cells and can occur in the sacrococcygeal area, ovaries, testicles, or other sites in the body.[5,6] The overall incidence of germ cell tumors is lower in children than in adults, malignant subtypes such as seminoma and embryonal carcinoma are absent in prepubertal patients but common in adults and teratomas, which are uniformly benign in children, may be malignant in adults.[1,2]

Most patients with testicular teratoma present with a hard and painless scrotal mass, which frequently appears non-homogeneous solid mass, often with calcifications on

ultrasonography. Sometimes atypical presentation such as transilluminating scrotal mass, due to the presence of internal cystic areas, detectable at ultrasonography.[7,8] We report the case of a completely cystic testicular teratoma, mimicking a simple cyst, in an infant.

According to the Gonzalez-Crussi histopathological classification, there are three types of teratoma: MT, immature teratoma (IT) and malignant teratoma.[9] Both mature and immature components are benign, and their potential for metastasis is unclear. In view of the benign nature of teratomas, testis sparing surgery may be considered.[10]

CASE REPORT

A 5 months-old child presented with a history of right sided scrotal swelling noticed by mother during bathing. On examination, a firm, non-tender, non-reducible mass was palpated indistinguishable from the right testicle and demonstrated no change in size during crying or straining (Figure 1).

Mother had no significant antenatal history with all normal antenatal scans. This child was born full term and had no genito-urinary or bowel complaints. There was no prior family history of cancer or testis masses. Complete blood count and basic metabolic panel were within normal limits. A scrotal sonography was performed and revealed a large anechoic cystic lesion measuring 28×18 mm within the right scrotal sac. No septation, calcification, or solid component were seen. The left testicle appeared to be of normal size and shape. The serum levels of AFP and β-hCG were within the normal range for his age. β-hCG was<0.100 mIU/ml and AFP were 35 ng/ml (Ref. 6-1045 ng/mL).

Child was taken for the surgery and testis-sparing tumor enucleation was performed. The tunica albuginea was incised, and the cyst was carefully removed intact without rupture (Figure 2).

Enucleated cystic mass was sent for histopathological examination. Gross specimen showed a unilocular cystic mass measuring 2.5×2.0×1.5 cm. Cut surface revealed cystic areas filled with keratinous material, with focal areas of cartilage and mucoid substance. No hemorrhage or necrosis noted. Microscopic examination showed flat to stratified epithelium with keratinization, hair follicular differentiation with scattered intestinal-type glands with goblet cells (Figure 3). No evidence of GCNIS or no cytologic atypia. A diagnosis of prepubertal-type MT was thus made, and the surgical excision was considered adequate and sufficient.

The post-operative period was uneventful; the patient recovered well and was discharged after 48 hrs. child was kept under observation in follow-up and repeat USG scrotum after 2 months revealed a normal-sized right testis.

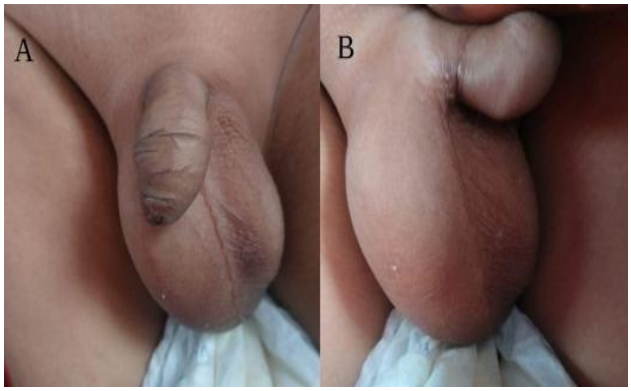

Figure 1 (A and B): Right sided scrotal swelling-firm, non-tender, non-reducible swelling in right hemiscrotum indistinguishable from the right testicle and demonstrated no change in size during crying or straining.

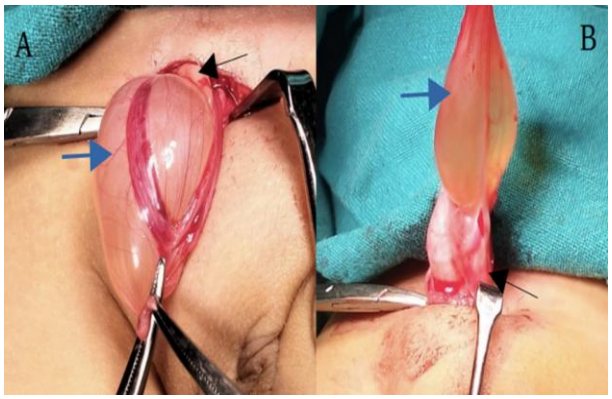

Figure 2 (A and B): Tense cystic mass measuring 28×18 mm, in close association with right sided testis and without any solid component (marked with blue arrow). Right sided testis (marked with black arrow) appears normal and the cyst was carefully removed intact without rupture.

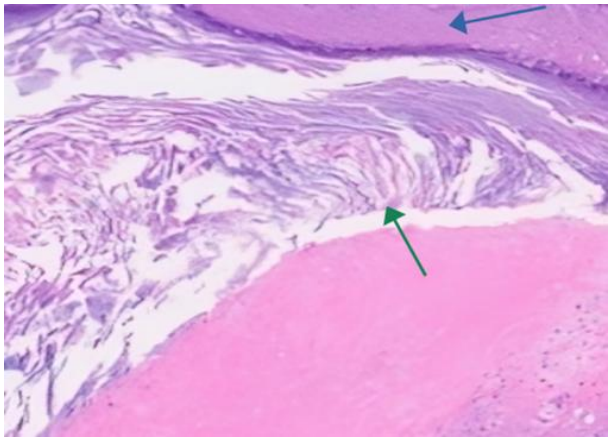

Figure 3: Flat to stratified epithelium with keratinization (marked by blue arrow), hair follicular differentiation (marked by green arrow showing flaky keratinous material) with scattered intestinal-type glands with goblet cells.

DISCUSSION

Testicular teratoma is a uniformly benign entity in prepubertal children and have high potential of being malignant in adults.[1-3,5] These are complex tumors derived from all three germ layers (endoderm, mesoderm and ectoderm). Prepubertal-type teratomas are those non-associated with GCNIS; they do not have significant cellular atypia and no metastasis. They are divided into mature (contain exclusively adult cells) and immature (contain embryonic or fetal cells).[4,5] This benign course of the tumor has encouraged the acceptance of a testis preserving approach to treatment. While Altadonna et al reported testis preserving surgery as a definitive treatment option for excision of benign testicular cysts.[11] Weissbach first applied this approach to 2 patients with teratoma.[12] Rushton et al provided extensive pathological analysis of 5 teratoma specimens removed by testis sparing procedure to ascertain intrinsic safety of this approach.[13]

Painless scrotal mass is the most frequent clinical presentation. Tumour markers like AFP, beta-human gonadotropin chorionic may contribute to the diagnosis and management of a testicular mass in boys. Ultrasonography is the best imaging modality to study testicular tumors. A benign tumour is suggested when USG shows mainly cystic component, well-defined borders, echogenic rim/normal to increased echogenicity lesion when compared to healthy testicular parenchyma. Malignant tumour is suspected when ultrasonography shows inhomogeneous, hypoechoic, not well-circumscribed or diffuse infiltration lesion.[7,14] In most patients, testicular teratoma is hard mass; however, it may sometimes have cystic quality on palpation because a part of the mass is composed of cysts filled with fluid or mucous. There are only handful of reports of patients with unilocular, completely cystic teratoma to date.

Testicular prepubertal-type MT in the prepubertal age group is typically found in children <5 years of age.[15] Typically, prepubertal-type MT manifests as a well-defined, rounded mass with cystic characteristics. The AFP in the serum is released by yolk sac cells during early fetal development, as well as by the proximal small intestine and the liver. Its level typically stays elevated, and it only normalizes around the age of 9 months. In the present case, the levels of AFP and β-hCG were normal. With these characteristics and USG findings diagnosis of mature teratoma was made. Surgery has been extensively utilized to treat benign testicular tumors in children, such as prepubertal-type MT. The prognosis of testis sparing surgical excision of tumor is excellent during childhood; it may not increase the possibility of recurrence in cases of benign tumors and is considered safe and feasible.[5]

CONCLUSION

In conclusion, early diagnosis and surgical intervention are crucial for excellent outcomes with a favorable prognosis and minimal risk of recurrence. Serum markers, such as AFP and HCG, and ultrasonography are first-level examinations, leading the subsequent management. Indeed, testis-sparing surgery is recommended in prepubertal age patients with a testicular mass with a benign ultrasonographic aspect and negative serum markers.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Jain N, Agrawal A. Prepubertal-type mature cystic teratoma of the testis in an infant: a rare benign entity with excellent prognosis-managed with testis sparing approach. Int J Contemp Pediatr 2025;12:1854-7.